

# Case report of pulmonary endometriosis and review of the literature

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## Abstract

Pulmonary endometriosis is a rare form of thoracic endometriosis. We herein describe a 29-year-old woman with recurrent hemoptysis associated with her menstrual cycle. The patient had a 4-month history of catamenial hemoptysis without thoracic pain, respiratory embarrassment, cough, fever, night sweating, or loss of appetite. Chest computed tomography revealed exudation shadows in the right lower pulmonary lobe and small fiber lesions in the right middle lobe and left lung. Thoracoscopic wedge resection of the right lower pulmonary lobe was performed, and the pathological result was pulmonary endometriosis. No evidence of hemoptysis during menstruation was found following the operation.

## Keywords

Pulmonary endometriosis, catamenial hemoptysis, menstrual cycle, recurrent, pathology, computed tomography, thoracoscopic wedge resection

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## Introduction

Endometriosis is characterized by growth of the endometrium outside the uterine cavity and is associated with infertility, dysmenorrhea, menorrhagia, and chronic pelvic pain. The incidence of endometriosis is 15% among reproductive-age women.<sup>1,2</sup> Thoracic endometriosis is a form of endometriosis that can be classified as either pleural or pulmonary. Pulmonary

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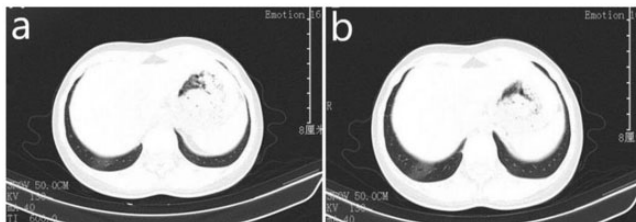
endometriosis was first described by Schwarz in 1938.<sup>3</sup> It is an extremely rare disease with complex causes and pathogenesises that remain unclear. In this report, we present a case of pulmonary endometriosis manifesting as recurrent hemoptysis associated with the menstrual cycle. This case is being reported to alert clinicians to the fact that pulmonary endometriosis can be readily misdiagnosed as a systemic disease because of its clinical manifestations and imaging signs. This case provides a reference for diagnosis and treatment of pulmonary endometriosis.

### Case report

A 29-year-old Chinese woman was referred to the outpatient department of our hospital on 1 September 2016 with a 4-month history of catamenial hemoptysis. The patient also had a history of a laparoscopic operation for treatment of an ectopic pregnancy and a 6-year history of secondary infertility.

In May 2016, the patient developed her first episode of catamenial hemoptysis with no prodrome. During the following 4 months, the hemoptysis became associated with her menstrual cycle. The patient reported that she began coughing blood on the first or second day of her menses and produced about 150 mL of hemoptysis during her menstrual cycle. She had no thoracic pain, respiratory embarrassment, cough, fever, night sweats, or weight loss.

On 1 September 2016, chest computed tomography (CT) revealed exudation shadows in the right lower pulmonary lobe (Figure 1(a)) and small fiber lesions in the right middle lobe and left lung (Figure 1 (b)), which were suggestive of pneumonia and endometriosis. The patient's clinical symptom returned when menstruation began, but she did not receive treatment. The patient agreed to undergo an examination on 27 September 2016 to obtain a definitive diagnosis. Chest CT revealed patchy infiltration in the right lower pulmonary lobe (Figure 2(a)) and small fiber lesions in right middle lobe (Figure 2(b)) and left upper lobe (Figure 2(c)); the clinical diagnosis was pulmonary endometriosis. Thoracoscopic wedge resection of the right lower pulmonary lobe was performed on 30 September 2016. We observed slight pleural adhesion under thoracoscopy; local stasis of blood in the lower lobe of the right lung, which was soft and had a smooth surface; no blood stasis in the right middle and lower lobes, chest wall, and pericardium; and no hydrothorax in the right thoracic cavity. The pathologic examination revealed chronic inflammation with hemorrhage and hemosiderin deposition in the lung tissue. Immunohistochemistry examination revealed estrogen receptor negativity, progesterone receptor negativity, CD positivity in alveolar epithelial cells, p53 negativity, and Ki67 positivity in some cells. All of these results were consistent with endometriosis. No recurrence or



**Figure 1.** Chest computed tomography revealed (a) exudation shadows in the right lower pulmonary lobe and (b) small fiber lesions in right middle lobe and left lung.



**Figure 2.** Chest computed tomography revealed (a) patchy infiltration in the right lower pulmonary lobe as well as small fiber lesions in the (b) right middle lobe and (c) left upper lobe.

complications were observed at the 4-month follow-up.

## Discussion

Endometriosis is characterized by growth of the endometrium outside the uterine cavity. Its incidence is 15% among reproductive-age women, and 32% to 48% of affected patients develop infertility or chronic pelvic pain. Endometriosis mostly occurs in the ovaries, followed by the rectouterine pouch and uterosacral ligament. It also occurs in the bladder, ureter, intestines, appendix, navel, and groin but is rarely seen in the lung, pleura, and kidney. The rate of malignant transformation of endometriosis is 1%.<sup>4</sup> Pulmonary endometriosis has multifactorial causes, but the disease pathogenesis remains unclear. Various theories have been proposed, including coelomic metaplasia, retrograde menstruation, stem cell theory, and microembolization theory. Most scholars have considered that pulmonary endometriosis is caused by peritoneal implants that make their way through fenestrations in the diaphragm to seed the pleural space, and a previous study demonstrated that these fenestrations seem to be present only on the right side of the diaphragm.<sup>5</sup> Other research has shown that the endometrium invades the interstitium of the lung by hematogenous or lymphatic metastasis during pelvic operations or

delivery. However, these theories fail to clearly state the mechanism of pulmonary endometriosis.<sup>6</sup>

Pulmonary endometriosis usually manifests as catamenial hemoptysis and always occurs at the onset of menses. The hemoptysis also stops after the cessation of menstruation and may or may not be accompanied by chronic cough, catamenial pneumothorax, catamenial hemothorax, recurrent episodes of low-grade fever, and asymptomatic lung nodules. The pathological mechanism of hemoptysis is based on the theory that estrogen can maintain the continuity of the vascular endothelium or increase the blood coagulability. It has been shown that high doses of estrogens reduce bleeding from the vascular malformations in patients with hereditary hemorrhagic telangiectasia and that this effect may be due to the ability of estrogens to maintain the continuity of the vascular endothelium or by their effect on blood coagulation.<sup>7</sup>

No definitive guidelines for the diagnosis and treatment of pulmonary endometriosis have yet been established. It is generally agreed that this condition affects women of reproductive age with a history of pelvic surgery, childbirth, or an operation involving the uterine cavity; hemoptysis occurs in association with the menstrual cycle along with catamenial hemothorax or chest pain with chronic cough; the

hemoptysis generally occurs before, after, or during menstruation, and the duration of symptoms varies; and lung CT reveals characteristic features of radiological examination of pulmonary endometriosis, namely pulmonary effusion or nodules with homogeneous density, clear boundaries, and appearance during the menstrual period with disappearance during the period of time between two menstruations. The clinical manifestations and pathologic features of the patient in the present case are consistent with the diagnosis of pulmonary endometriosis.

The primary pathological basis of pulmonary endometriosis is periodic shedding and bleeding of the endometrium. Furthermore, blood invades the interstitium of the lung and spreads to the surrounding area, leading to exudative change. Absorption and clearance of the hemorrhagic lesions then occurs between two menstruations. In the present case, long-term incomplete absorption or discharge of the hemorrhagic lesions led to hemosiderosis deposition, chronic inflammation, and fibroplasia. The pathological diagnosis was pulmonary endometriosis. However, according to the literature, histological changes are not always specific for a pathological diagnosis of pulmonary endometriosis, and such evidence can only be found in 30% of patients.<sup>8</sup> In addition, misdiagnosis of pulmonary endometriosis may readily occur due to the non-specificity of X-ray and CT manifestations. Chest CT can be used as a supplementary technique in the diagnosis of pulmonary endometriosis. Because of its clinical manifestations and imaging findings, pulmonary endometriosis can easily be misdiagnosed, and other diseases must be ruled out.

Therapy for pulmonary endometriosis includes conservative treatment and surgery, and the most appropriate therapy is

chosen according to the patient's clinical symptoms, severity, and requirement. Gonadotropin-releasing hormone agonists are widely accepted as the first-choice drugs; these agents inhibit ovarian release of estrogen and progesterone, leading to reduced levels of plasma estradiol. This achieves a state of pseudo-menopause and inhibition of lesion growth. The shortcomings of these drugs are their high rate of adverse effects and their ability to alleviate symptoms in a short time but without thorough removal of the lesions. In addition, the cost of prolonged treatment and the long-term recurrence rate are high, and the drug may even affect pregnancy. After failure of drug treatment of the development of intolerable adverse effects, most patients choose operative treatment methods. The routine operative technique is total pneumonectomy or pulmonary wedge resection, which have a lower recurrence rate and good outcomes compared with drugs and thoracoscopic lobectomy.<sup>9</sup> According to the literature, a small percentage of patients who undergo no treatment can experience an easing of symptoms or spontaneous resolution of the endometriosis.<sup>10</sup>

### **Ethics and consent statements**

This study was conducted in accordance with the declaration of Helsinki and with approval from the Ethics Committee of Hongqi Affiliated Hospital of Mudanjiang Medical College. Written informed consent was obtained from the patient.

### **Declaration of conflicting interest**

The authors declare that there is no conflict of interest.

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